Nasopalatine duct cyst mimicking as residual cyst- An enigma in clinical diagnosis- A case report

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ABSTRACT

Incisive canal cyst or nasopalatine duct cysts (NPDCs) are the most common non-odontogenic cysts of the maxilla. These are the common intraosseous developmental cyst occurring in the midline of the maxillary anterior region. They are usually asymptomatic and are discovered incidentally during the routine radiographic examination. They can be described as a well-circumscribed radiolucency between the root apices of the maxillary central incisors. Since these lesions show diagnostic dilemma in clinical and radiological study, a definitive diagnosis is required to be confirmed by histopathology. Surgical approach with enucleation of the cyst is the definitive treatment of NPDC. Hereby, we report an unusual presentation of NPDCs mimicking as residual cyst which was associated with missing upper anterior teeth.

Key words: *Cyst, Maxilla, Nasopalatine cyst, Palate, Radicular cyst*

The nasopalatine duct cyst (NPDC) was first described by Meyer in 1914 [1]. Recently, the World Health Organization has classified it as a non-odontogenic, developmental, and epithelial cyst of the maxilla which is believed to arise from the remnants of the nasopalatine duct. Most of these cysts develop in the midline of anterior maxilla, just behind the root apices of the maxillary central incisors which may cause common confusion with the periapical lesions. Incidence is 1.7–11.9% with a slight male predilection [2]. Many NPDCs are diagnosed during routine periapical or occlusal radiographs. Here, we report a case of NDPC mimicking residual cyst.

CASE REPORT

A 35-year-old adult female patient reported to the department of oral medicine and radiology with the chief complaint of pain in the upper front teeth region for 1 week. Pain was insidious in onset, intermittent, throbbing type which got aggravated on chewing and lying down posture. The patient gave the history of extraction of her upper front teeth 1 month back due to caries and got it replaced 20 days back. Her medical history was non-contributory. Extraoral examination showed no abnormality, and no lymphadenopathy was detected.

Intraoral soft tissue examination revealed suture scar in alveolar mucosa just above the mucogingival junction in relation to teeth no. 21, 22, and 23 (Fig. 1). On inspection, a diffuse swelling was seen confined to the upper labial vestibule in relation to teeth no. 21 and 22 measuring approximately 1 × 1 cm extending mediolaterally from the labial frenum to distal aspect of 22, superoinferiorly from mucogingival junction to obliterate the labial vestibule. The overlying mucosa appeared normal. On palpation, swelling was tender and soft in consistency. Hard tissue examination revealed metal ceramic bridge in relation to teeth no. 11, 12, 21, 22, and 23. Based on the history and clinical examination, a provisional diagnosis of residual cyst in relation to teeth no. 21 and 22 was arrived. NPDC, large incisive canal, and odontogenic keratocysts were considered under differential diagnosis.

An intraoral periapical radiograph was advised which revealed missing teeth in relation to 11, 21, and 22 and a solitary well-defined ovoid radiolucency in the periapical region of edentulous region measuring approximately 2.5 × 1.5 cm with a well-corticated border. It extended from 0.5 cm away from the alveolar crest inferiorly to the floor of the nasal fossa superiorly. Mediolaterally, it extended 0.5 cm and 1cm away from the midline on the left and right side, respectively. The internal structure was completely radiolucent (Fig. 2).

Routine hematological investigations showed no abnormality and fine-needle aspiration of the swelling showed clear straw-colored fluid suggesting inflammatory lesion. Enucleation of the cyst was done under local anesthesia, and the specimen was sent for histopathological examination (Fig. 3). The histopathology report revealed cystic lumen lined by an epithelium and an underlying connective tissue capsule. The epithelium was 2–3 layers in thickness and varied from flat to cuboidal to columnar. The

epithelium-connective tissue interface was flat. The underlying connective tissue showed dense collagenous component and mild chronic inflammatory infiltrate predominantly comprising of lymphocytes. Numerous blood vessels lined by endothelial cells were seen with extravasated red blood cells. No evidence of odontogenic epithelium was seen (Fig. 4). These histopathological features were suggestive of NPDC. The patient was recalled post-operatively, after 4 months following enucleation. There was no recurrence, and follow-up is still going on (Fig. 5).

DISCUSSION

The NDPCs are the most common developmental, epithelial, and non-odontogenic cysts of the maxilla which are derived from the proliferation of embryonic epithelial remnants of the nasopalatine duct [2]. Most of the cases occur in the 4th–6th decades of life with male predilection. However, in our case, middle-aged female was the victim. Triggering factors include infection (38%), trauma (16%), minor salivary mucus retention, and inflammatory stimulus [3]. Most of these cysts are asymptomatic or cause minor symptoms. It presents as a small, well-defined swelling posterior to the palatine papilla. The swelling is usually fluctuant, and the deeper layers are covered by normal-appearing mucosa unless ulcerated from masticatory trauma. If the cyst expands, it may penetrate the labial cortical plate and produce a swelling inferior to the maxillary labial frenum. Pressure from the cyst on the adjacent nasopalatine nerves may cause a burning sensation or numbness over the palatal mucosa [4]. Many NPDCs were diagnosed during routine periapical or occlusal radiographs. In the present case, the patient was presented with a pain and diffuses swelling in the labial vestibule.

Radiographically, NPDC appeared as a well-defined, round, or oval radiolucency located between the root apices of maxillary central incisors with sclerotic border. Superimposition of anterior nasal spine gives the lesion typical heart-shaped radiolucency [5]. Our case presented with a ovoid radiolucency in the anterior maxilla in the region of missing teeth no. 21, 22, and 11. NPDC can be differentiated from other radiolucent lesions in maxillary

Figure 1: Suture scar with diffuse swelling in the upper labial vestibule in relation to 21, 22, and 23

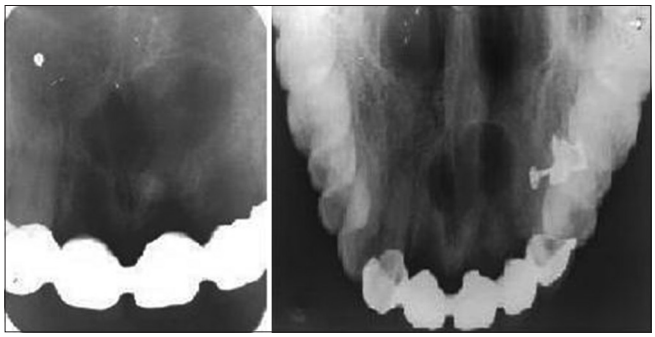

Figure 2: Intraoral radiographs revealed missing 11, 21, and 22 and ovoid radiolucency at the periapical region of 21 and 22 (arrow) measuring approximately 2.5 × 1.5 cm surrounded by a corticated border

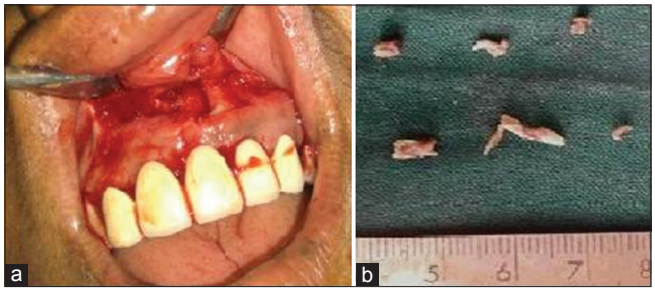

Figure 3: (a) Surgical enucleation of the cyst done through labial approach and (b) enucleated specimen

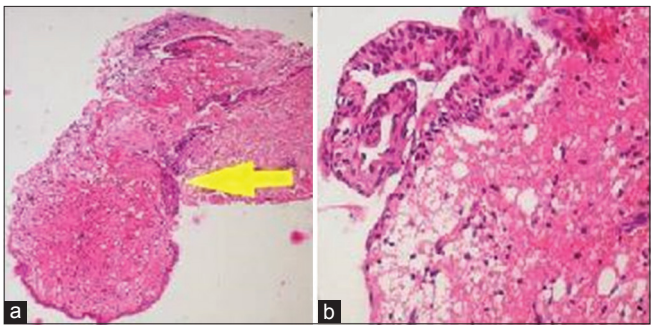

Figure 4: (a and b) Photomicrograph of H and E stained sections showing stratified squamous epithelium of variable thickness indicated by arrow and underlying connective tissue with dense chronic inflammatory cell infiltration and numerous blood vessels

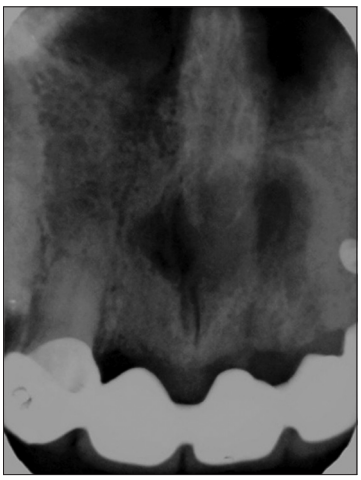

Figure 5: Intraoral periapical radiograph taken at 4-month follow-up showing bone fill at the alveolar defect

midline, such as large incisive foramen, lateral radicular cyst, or lateral periodontal cyst. A foramen larger than 6 mm may simulate the appearance of a cyst. Distinction of NPDC from a large incisive fossa can be made clinically by aspiration of the lesion. A radicular cyst is usually associated with non-vital teeth and shows loss of continuity of the laminadura while the NPDC is usually associated with vital teeth. In this case, although the tooth was missing, a second periapical view taken at a different horizontal angulation which showed an altered position of the radiolucency, whereas radicular cyst is always centered at the root apex of the tooth [4,5].

Histologically, NPDC lesion is characterized by epithelial lining surrounded by connective tissue wall with varying degree of inflammation and presence of nerve bundles, mucous glands, and adipose tissue. The epithelial lining of the cyst is variable ranging from squamous, columnar, cuboidal, or combination of these epithelium [5]. These features were correlating with the present case, and there was no evidence of odontogenic epithelium, thereby ruling out the possibility of residual cyst. The aggressive nature of these cysts should not be underestimated as they can cause massive destruction and disability. Further, squamous cell carcinoma in the maxilla originating from NPDC can also be considered as differential diagnosis [6].

Asymptomatic small NPDC does not require any treatment [6,7]. If the cyst is infected or shows progressive enlargement, then surgical intervention is carried out preferably from the palate to avoid damage to the nasopalatine neurovascular bundle. Recurrence after surgical excision is very rare [8]. In the present case, the lesion was localized over labial vestibule, and thus, enucleation was performed via labial approach.

CONCLUSION

Although NPDCs are the most common non-odontogenic cysts, it can mimic any odontogenic cysts and tumors in the anterior maxilla, especially when associated with missing teeth. As a specialist in oral diagnosis, a thorough knowledge of this cyst is essential to give a definite diagnosis along with clinico-radiographical correlation.

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